

GIANT CARVENOUS HEMANGIOMA OF THE ANTERIOR CHEST WALL IN A NEONATE: A CASE REPORT

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SUMMARY

A term, Malay male neonate, delivered by spontaneous vertex delivery, was found to have a huge carvenous hemangioma on the anterior chest wall. He developed respiratory failure on the second day of life. Having considered the various options possible, an emergency surgical excision was carried out at the age of 57 hours. The patient recovered after a stormy post-operative period.

INTRODUCTION

Carvenous hemangioma is a common benign neoplasm in the paediatric age group. It can occur in any part of the body. However, carvenous hemangioma, especially of the 'giant' variety, arising from the anterior chest wall has only rarely been reported.

There is no strict definition for the word 'giant' when used with hemangioma. In most case reports,

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tumour size of at least 5-6 cm in diameter is said to be 'giant'.¹ We report here a case of giant carvenous hemangioma, 25 cm in diameter, arising from the anterior chest wall which was treated by excision.

CASE HISTORY

A Malay, male infant was born at term gestation by spontaneous vertex delivery to a 22-year-old primigravida with uneventful pregnancy. His birth weight was 3.75 kg. Apgar score at one minute after birth was nine. At birth, a large swelling was found over his anterior chest wall.

He was referred to the paediatric surgeon on the second day of life.

On examination, he was afebrile and mildly jaundiced. He was pink in air; heart rate was 150/min; blood pressure 96/67 mmHg; respiratory rate 70/min. There were bilateral conjunctival hemorrhages. There was no petechie on the skin.

Extending across the anterior chest wall was a soft, spongy, bluish-red swelling, measuring 25x20x10 cm. It had a very broad base. No bruits were heard over the swelling which was non-pulsatile (Fig. 1). Dilated veins were present over the rest of the anterior chest wall and abdominal wall. The direction of blood flow was towards the swelling. The apex of the heart could not be palpated. The heart sound was dual rhythm



Fig. 1 Before operation, appearance of patient with hemangioma over the anterior chest wall.

and there was no murmur. The breath sounds were vesicular and equal in both lungs. The liver was 2.5 cm palpable below the right costal margin at the mid clavicular line. Other systems were normal.

Investigations done on admission: Hb: 14.5 gm%; TWBC: 11,000/cu mm (polymorphs 58%, lymphocytes 42%); platelets 90,000/cu mm; blood urea and serum electrolytes were normal; random blood sugar was 4.2 mmol/l; total serum bilirubin was 124mmol/l; G6PD screening was normal.

Both the mother and the baby's blood groups were O Rhesus positive. Blood culture yielded no growth. AP view of the chest X-ray showed a large circular soft tissue swelling over the chest extending beyond the edge of the thoracic cage. The cardiac border could not be delineated.

The lateral view of the chest X-ray (Fig. 2) showed soft tissue swelling arising from the anterior chest wall but separate from the heart. Electrocardiogram could not be obtained because of the presence of the mass.

Progress of the patient

The patient became progressively cyanosed over a period of 12 hours. The heart rate increased to 164/min, respiratory rate 100/min. He was given 100% oxygen *via* headbox which did not completely relieve the cyanosis. The liver was palpable 3 cm below the right costal margin. There was no apparent enlargement of the mass.

An emergency operation was carried out at the age of 57 hours. The large spongy swelling with broad base was found to have numerous feeding vessels arising from the intercostal vessels. Total excision was carried out. The mass weighed 150g. About 1000 ml of blood was lost during operation. This included the amount estimated to be sequestered in the mass. The blood loss was replaced by 750 mls of whole blood and 250 mls of fresh frozen plasma. Primary skin closure was

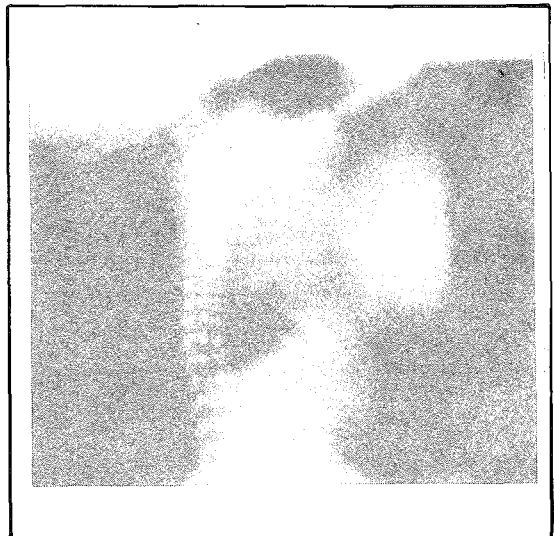


Fig. 2 Lateral roentgenogram of chest taken before operation showing large soft tissue swelling arising from the anterior chest wall.

established after undermining the surrounding skin (Fig. 3).

Histopathological findings in the excised mass were consistent with the diagnosis of giant carvenous hemangioma.

Post-operatively, the patient turned cyanosed after extubation despite the ability to breathe spontaneously. He was reintubated and was put on IPPV with FiO₂ of 60%. Chest X-ray showed cardiomegaly with pulmonary vascular engorgement. His fluid intake was restricted and intravenous frusemide was administered. His blood gases were satisfactory. Repeat blood count showed: Hb 9 gm%; TWBC 5,400/cu mm (polymorphs 68%, lymphocytes 32%) and platelets 100,000/cu mm.

14 hours post-operation, however, he developed generalised cyanosis. His heart rate was 190/min

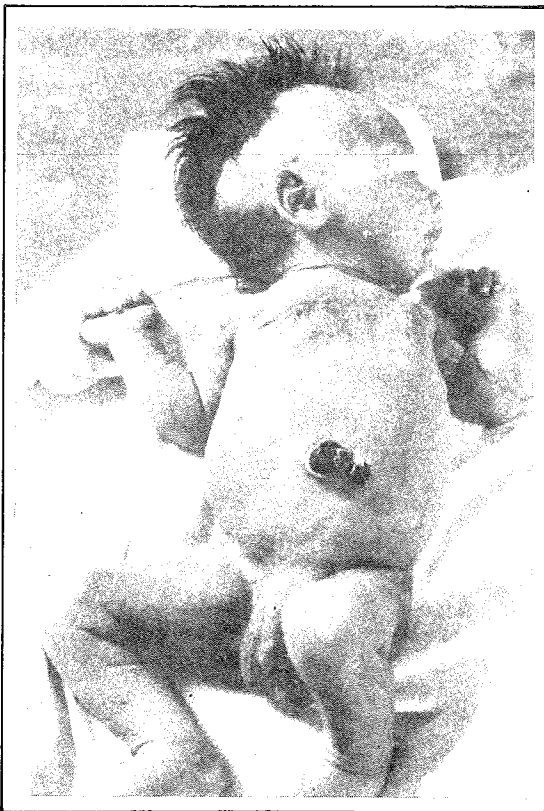


Fig. 3 Long surgical scar extends across anterior chest wall of the patient postoperation.

and blood pressure was 64/49mm Hg. There was triple rhythm in the heart sounds. Abdominal distension was marked and liver size could not be assessed.

Treatment with digoxin and frusemide was started. Cyanosis abated within 30 minutes after intravenous injection of both drugs. His heart rate was 159/min three hours later. Intermittently, he turned cyanosed whenever he was restless. He was sedated with intravenous morphine which enabled him to be weaned off the ventilator support by the age of 16 days. Digoxin was stopped at age of 24 days. Chest X-ray repeated showed heart size to be within normal limits.

He was discharged at the age of 32 days.

DISCUSSION

The rapid onset of tachypnea followed by cyanosis in this patient after birth heralded the presence of respiratory failure and possibly cardiac failure as well. Before birth, oxygenation of the baby relied solely on the placenta. At birth, with the clamping of the umbilical cord, tissue oxygenation depended on the efficiency of the patient's own cardiopulmonary function. In this patient, two aggravating factors were evident: firstly the weight of the giant cavernous hemangioma compressing the chest wall thus affecting its compliance and leading to respiratory insufficiency, and secondly arteriovenous shunts within the hemangioma giving rise to increased circulatory blood volume which resulted in cardiac decompensation. The presence of the latter has been described by Cooper *et al.*² These shunts provided 'low' pressure circuit through which passed a large part of the left heart output with an initial left diastolic overload. This was followed by compensatory cardiac dilatation and hypertrophy, eventual overloading of right heart and consequent cardiac failure.³

The occurrence of cardiac failure post-operatively in this patient was not an unexpected phenomenon.³ The interruption of large number of A-V shunts by surgical removal of the

hemangioma caused the high circulating blood volume to come against the 'high' systemic vascular resistance, increasing the systolic load on the left side of the heart that previously had only the diastolic overload placed on it by the blood return through the A-V shunts in the hemangioma. This increased the strain on the heart and precipitated failure.

Although thrombocytopenia has been reported as a complication of large and/or multiple hemangiomas, this was not marked in this patient. This could be explained by the fact that the hemangioma was removed shortly (less than 72 hours) after birth before platelet trapping could occur in significant number. In reports where thrombocytopenia was present, the patients presented at an older age.^{4,5,6}

Various modes of treatment for giant carvenous hemangioma have been advocated:^{4,7} sclerotherapy, irradiation, steroid therapy and surgical excision.

Sclerotherapy has been used in some patients with good results.⁸ Immediately after sclerotherapy, however, the hygroscopic action of the sclerosing fluid caused further swelling of the hemangioma. In our patient, this unavoidable reaction would have been disastrous.

Radiotherapy was not considered at all in this patient. Apart from radiation hazards to the surrounding organs and tissues,⁹ at least three weeks of irradiation is required to initiate resolution of the hemangioma.⁶

Studies on adrenalectomised rats have shown that corticosteroids increased vascular sensitivity to circulating vasoconstrictive agents.¹⁰ The use of steroid for the successful induction of regression of rapidly growing and large cavernous hemangioma has been reported by various authors since 1967.^{7,11} Again, however, at least one week of therapy is needed to bring about the onset of regression of the hemangioma.¹¹

In our patient, the most pressing problem was respiratory failure caused directly by the hemangioma which required immediate relief. Most authors would agree that surgery would be the treatment of choice in such situations.¹¹

Surgery was made relatively easy in this patient by the absence of involvement of the underlying organs. The latter was assessed clinically by the relative mobility of the hemangioma over the chest wall and by plain X-ray appearance. Angiographic studies, although desirable, could not be performed due to the rapid deterioration of our patient. The hemangioma had a short pedicle around which a tourniquet was applied prior to commencement of excision. Nevertheless, the blood loss was still considerable despite excessive use of coagulation diathermy. The problem of volume replacement was compounded by the fact that tachycardia continued unabated throughout surgery and in the immediate post-operative period making assessment difficult. Although the hazards of surgery in such cases are prohibitive, the risks have to be accepted in this patient since no other options were available. This high risk procedure would not have been attempted without the presence of good anaesthetic support and intensive care facilities.

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